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Breathing retraining - A five-year follow-up of patients with dysfunctional breathing

Carina Hagman [a,b,c,*], Christer Janson [b], Margareta Emtner [b,c]

[a] Department of Physiotherapy, Falu Hospital, SE-791 82 Falun, Sweden
[b] Department of Medical Sciences, Respiratory Medicine and Allergology, Uppsala University, SE-751 85 Uppsala, Sweden
[c] Department of Neuroscience, Physiotherapy, Uppsala University, Box 593, SE-751 24 Uppsala, Sweden

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Summary
Introduction: The term dysfunctional breathing (DB) has been introduced to describe patients who display divergent breathing patterns and have breathing problems that cannot be attributed to a specific medical diagnosis. Patients with DB are often misdiagnosed as having asthma.
Objectives: To describe patients with DB, five years after a breathing retraining intervention.
Methods: Out of initially 25 patients with DB and 25 age and sex-matched patients with asthma, 22 patients with DB and 23 patients with asthma (ages 25–78 years) were followed up after five years. Data were collected from posted self-report questionnaires. Only patients with DB had received breathing retraining, consisting of information, advice and diaphragmatic breathing. Patients were evaluated regarding quality of life (SF-36), anxiety, depression, sense of coherence, hyperventilation, influence on daily life, emergency room (ER) visits, and symptoms associated with DB.
Results: Quality of life (SF-36), physical component summary scale (PCS), had improved in patients with DB from 43 to 47 ($p = 0.03$). The number of ER visits had decreased from 18 to 2 in patients with DB ($p = 0.02$). Symptoms associated with DB had decreased extensively, from a mean score of 6.9 to 2.7, on a DB criterion list ($p < 0.001$). Patients with DB were less impaired by their breathing problems both in daily life and when exercising ($p < 0.001$). The only difference found over time in the asthma group concerned quality of health, bodily pain, which had deteriorated, from 77 to 68 ($p = 0.049$).
Conclusion: This five-year follow-up study indicates that patients with dysfunctional breathing benefit from breathing retraining.

* Corresponding author. Department of Physiotherapy, Falu Hospital, SE-791 82 Falun, Sweden. Tel.: +46 23490895; fax: +46 23490669.
E-mail address: carina.hagman@ltdalarna.se (C. Hagman).

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Introduction

Studies indicate that as many as one-third of individuals with physician-diagnosed asthma in developed countries, are overdiagnosed and do not actually have asthma.[1,2] Markund claims that about 10% of patients diagnosed as having asthma instead suffer from "functional breathing disorder".[2] The term dysfunctional breathing (DB) has been introduced to describe patients who display divergent breathing patterns[3] and have breathing problems that cannot be attributed to a specific medical diagnosis such as asthma, chronic obstructive pulmonary disease or sensory hyper-reactivity.[4] Dysfunctional breathing can be associated with symptoms that may be wrongly attributed to asthma,[5] and individuals with dysfunctional breathing often remain undiagnosed or are misdiagnosed as having asthma.[2,4,6] Incorrect treatment due to misdiagnosis will not only have consequences for the patients, but will also have consequences for health economics, as asthma medications are often prescribed to patients with breathing problems but without asthma.[2] DB can also occur in association with genuine asthma and other airway diseases[7] and there is a risk that these patients are prescribed excessive doses of asthma medication.[6]

There is no consensus about a definition of or a diagnostic test for DB.[3,4] Van Dixhorn suggests that DB comprises a wider aspect of respiratory function than hyperventilation, including breathing pattern and body awareness.[8] Thomas has shown that patients treated for asthma and with symptoms suggestive of dysfunctional breathing benefit from physiotherapy-based breathing retraining with improvements in quality of life and regression of symptoms.[9] The intervention included explanation of symptoms attributable to abnormal breathing and diaphragmatic breathing exercises.[9] Hornsveld also claims that many patients with abnormal breathing patterns benefit from breathing retraining.[10]

In a previous descriptive baseline study 25 patients with DB and 25 patients with well-controlled asthma were investigated.[11] The results indicated that patients with DB were more disabled because of their condition than patients with well-controlled asthma. They had poorer health-related quality of life, were more anxious, had poorer sense of coherence, higher prevalence of suspected hyperventilation, more breathing problems and their breathing problems were more negatively affected by stress than patients with well-controlled asthma. In addition, they had had breathing problem for a long time (mean seven years) before being correctly diagnosed, had often made emergency room visits and had previously been treated with ineffective asthma medication.

The aim of this follow-up study was to describe patients with DB who had received breathing retraining interventions, and were thereafter followed for five years. Describe possible changes in health-related quality of life, anxiety, depression, sense of coherence, hyperventilation, influence on daily life, asthma medication, number of emergency room visits and symptoms associated with DB. The asthma group was included as a control group and evaluated at baseline and at follow-up, as individuals with DB often are misdiagnosed as having asthma.

Methods

Subjects

In the baseline study, 25 patients who fulfilled the criteria for dysfunctional breathing (see below), but without a diagnosis of asthma, and 25 age and sex-matched patients with well-controlled asthma and no dysfunctional breathing were examined, included and evaluated at the Lung and Allergy Outpatient Department in Falun, Sweden.[11] The 50 patients fulfilled the following criteria: (i) 16–80 years; (ii) forced expiratory volume in 1 s (FEV_1) and vital capacity (VC) $\geq$ 80% of predicted value; (iii) resting oxygen saturation $\geq$ 95%; and (iv) no concomitant disease. All patients had been non-smokers for at least one year prior to the study. Patient characteristics at baseline are presented in Table 1.

The patients gave their informed consent prior to their inclusion in the study, and the study protocol was approved by the Ethics Committee of the Medical Faculty of Uppsala University.

Table 1 Patient characteristics at baseline (patients with data from baseline and follow-up). Number of patients (*n*) and mean (SD).

	DB ($n = 22$)	Asthma ($n = 23$)	*p*-value
Sex, F/M (n)	18/4	17/6	0.77
Age (yrs)	48 (13)	47 (14)	0.76
BMI (kg/m^2)	25 (3)	26 (3)	0.77
Smokers (n)	0	0	–
Ex-smokers (n)	5	6	1.0
Duration of the disorder/disease (yrs)	7 (8)	23 (13)	<0.001
Asthma medication (n)	14*	23	0.001
ER visits last 12 months (n)	8	4	0.19
Score of the 10-criterion list	7.6 (1.4)	1.3 (0.9)	<0.001

DB = dysfunctional breathing, BMI = body mass index, ER = emergency room, * before inclusion.

Identification of patients with dysfunctional breathing

Patients with breathing problems were examined by a physician, who confirmed that the patient did not have asthma or any other respiratory disease. As described previously,[11] the diagnosis of patients with DB was based on examination by a physician, the presence of a dominant high costal breathing pattern at rest (observed and assessed visually by the physician and the physiotherapist) and at least five out of ten symptoms: (i) difficult inspiratory breathing; (ii) unable to take deep breaths; (iii) increased breathing frequency (>16/min); (iv) frequent sighing/yawning; (v) frequent need to clear the throat; (vi) muscle and joint tenderness in the upper part of the chest (sternocostal joints and/or intercostal muscles): (vii) hacking cough; (viii) chest tightness; (ix) sensation of lump in the throat; and (x) previous or current effects of stress (*DB criterion list*).

Patients with asthma

Patients with well-controlled asthma had also been diagnosed and examined by a physician. The asthma diagnosis was based on patients' history, spirometry before and after bronchodilators and bronchial challenge testing. They were considered well-controlled if they were clinically stable, and if FEV_1 and VC were $\geq$80% of predicted value. All the patients with asthma used inhaled steroids, 19 patients had long-acting beta-2-agonists, 18 patients had short-acting beta-2-agonists, and 12 patients used oral steroids on demand. They were included in the study if they did not have a high costal breathing pattern at rest and fulfilled fewer than five symptoms from the DB criterion list.

Measurements

At inclusion, patients were investigated and answered five self-report questionnaires at the Department. After the patients had been examined by the physician, a physiotherapist examined them and asked about symptoms according to the DB criterion list. At the five-year follow-up, the same five questionnaires plus the DB criterion list were sent to the patients.

Medical Outcomes Survey Short Form 36 questionnaire (*SF-36*) measures general health-related quality of life. The SF-36 has 8 domains: physical function, role physical, bodily pain, general health, vitality, social function, role emotional and mental health. For each domain the score ranges from 0 to 100 (worst — best).[12] The first four domains constitute the physical component summary scale (PCS) and the last four the mental component summary scale (MCS).

Hospital Anxiety and Depression Scale (*HADS*) measures mental health,[13] yielding separate scores for anxiety and depression. The score range in HADS is 0—21, a higher score indicating poorer mental health.

Sense of Coherence (*SOC*). The 29 items (11 comprehensibility, 10 manageability and 8 meaningfulness) range from 29 to 203. The higher the score, the stronger the SOC.[14]

Nijmegens Symptoms Questionnaire (*NQ*) is suggestive as a measure of hyperventilation syndrome. The questionnaire assesses 16 symptoms associated with abnormal breathing on a five-point scale (0—4). A score of 24/64 or more is suggestive of a diagnosis of hyperventilation syndrome.[15,16]

Influence on daily life, a questionnaire consisting of items concerning breathing problems and effects of stress and impact on daily life and on exercise. Most of the answers were given on a visual analog scale (VAS) (0—10 cm, where 0 indicated the best value and 10 the worst). Most of the questions were related to the patient's experience of the past two months.

Questions regarding asthma medication and emergency room visits for breathing problems were also included.

DB criterion list. The criterion list used at baseline consisted of ten criteria, at follow-up the criterion "increased breathing frequency" was omitted.

Intervention

After inclusion and data collection at baseline, the patients with DB received one to four individual sessions of physiotherapist-supervised breathing retraining. The number of visits was based on the patients' needs. The first session lasted for 90 min and the following sessions for 60 min with 1—3 months in between. The sessions consisted of explanation of normal breathing, developing patients' awareness of their breathing patterns, and possible effects/symptoms of an upper chest breathing pattern, "dysfunctional breathing". They were taught diaphragmatic breathing in different body positions: supine, sitting and standing. They were encouraged to be aware of their breathing patterns and to practice breathing exercises several times each day, for example when they were lying in bed, sitting on a bus or watching television. In addition, the DB group also received information about their condition from the physician. The patients with asthma did not receive any physiotherapy intervention.

Statistical analysis

Analyses were undertaken with SPSS version 18.0. Data were analyzed for statistically significant differences between the groups using the unpaired *t*-test and Fisher's exact test. For differences within groups over time the paired *t*-test, McNemar test and Wilcoxon signed-rank test were used. Effects were considered significant at $p < 0.05$.

Results

Fifty patients answered the questionnaires at baseline and 45 (90%) at the five-year follow-up. Two patients, one in each group had died (one of sudden cardiac death and one of colon cancer) at the time of follow-up, and three patients (two in the DB group, one in the asthma group) did not return the questionnaires. Thus, data for 22 (88% of the original number) patients with DB and 23 (92%) patients with asthma were analyzed. Results include only patients who were available at baseline and at follow-up.

Health-related quality of life, and anxiety and depression

In health-related quality of life (SF-36), the domain "physical function" had improved in the DB group from 77 to 87 ($p = 0.04$) (Table 2). At baseline, the DB group had significantly lower values in "vitality", "social functioning", "role emotional" and "mental health" ($p < 0.05$) than the asthma group. At follow-up, there was no significant difference considering the domain "role emotional" between the two groups. The summary measure physical health (PCS) had improved in the DB group. The summary measure mental health (MCS) was lower in the DB group both at baseline and at follow-up than in the asthma group ($p < 0.05$) (Table 2). No changes were seen over time in either of the groups regarding the mean scores for anxiety and depression (Table 2). At baseline, 13 patients (59%) with DB had a HADS anxiety scores $\geq$8, indicating anxiety, as compared with 6 patients (26%) in the asthma group ($p = 0.04$). At follow-up there were 11 patients (50%) with DB, as compared with 3 patients (13%) in the asthma group ($p = 0.01$).

Sense of coherence and hyperventilation

Sense of coherence was stable over time in both groups (Table 2) with lower values in the DB group both at baseline and at follow-up ($p < 0.05$). There was a decreased mean score of the NQ in the DB group (Table 2). At baseline, 13 patients (59%) with DB had an NQ score $\geq$24, which indicates suspected hyperventilation, and at follow-up the result was nine patients (41%) ($p = 0.34$). The corresponding values (a score $\geq$24) for patients with asthma were four patients (17%) both at baseline and at follow-up.

Influence on daily life

Patients with DB had less breathing problems at follow-up ($p < 0.001$) (Table 3). Their breathing problems had less impact both on daily life and on exercise ($p < 0.001$), they were less often off work ($p = 0.02$), and their breathing problems were less affected by stress ($p = 0.03$). At baseline the DB group was more affected by stress than the asthma group ($p < 0.001$), but no significant group difference was found at follow-up. No differences in the prevalence of breathing problems were seen over time in the asthma group (Table 3).

Asthma medication, emergency room visits and symptoms associated with DB

Before inclusion 14 patients with DB were on asthma medication. None of the patients with DB had used asthma medication during the five-year follow-up. The asthma medication in the asthma group was virtually the same at baseline and at follow-up. At follow-up, the DB group had fewer emergency room visits due to breathing problems during the last 12 months, both regarding number of patients and number of visits. Number of DB patients with emergency room visits during the last 12 months had decreased from 8 to 2 ($p = 0.03$) and number of visits had decreased from 18 to 2 ($p = 0.02$) (Fig. 1). For the DB group, the mean score of the DB

Table 2 Results of health-related quality of life (SF-36), HADS anxiety (HADS-A), HADS depression (HADS-D), Sense of Coherence (SOC) and Nijmegens Symptoms Questionnaire (NQ). Number of patients (n) and mean (SD).

	DB_1 ($n = 22$)	DB_2 ($n = 22$)	p-value	$Asthma_1$ ($n = 23$)	$Asthma_2$ ($n = 23$)	p-value
SF-36						
PF	77 (21)	87 (17)	0.04	87 (14)	86 (17)	0.70
RP	64 (38)	72 (40)	0.40	74 (37)	85 (27)	0.06
BP	66 (27)	63 (25)	0.67	77 (29)	68 (26)	0.049
GH	54 (21)	62 (21)	0.12	62 (20)	65 (20)	0.41
VT	48 (22)	49 (25)	0.80	63 (23)	63 (19)	0.91
SF	70 (27)	71 (25)	0.87	93 (15)	88 (19)	0.09
RE	70 (44)	76 (39)	0.55	93 (25)	87 (24)	0.46
MH	72 (19)	68 (21)	0.36	82 (15)	82 (16)	0.96
PCS	43 (9)	47 (10)	0.03	46 (10)	47 (9)	0.74
MCS	43 (13)	42 (13)	0.69	52 (8)	59 (9)	0.50
HADS-A	9.3 (4.0)	7.8 (4.9)	0.08	5.2 (3.8)	4.5 (3.5)	0.25
HADS-D	6.0 (2.9)	5.0 (3.1)	0.12	3.6 (2.8)	3.4 (2.7)	0.55
SOC	134 (28)	136 (24)	0.65	155 (18)	160 (15)	0.10
NQ	27 (10)	22 (8)	0.03	15 (7)	16 (9)	0.91

DB = dysfunctional breathing, $_1$ = at baseline, $_2$ = at five-year follow-up.
PF = physical function, RP = role physical, BP = bodily pain, GH = general health, VT = vitality, SF = social function, RE = role emotional, MH = mental health.
PCS = physical component summary scale, MCS = mental component summary scale.
Low scores on SF-36 and SOC indicate greater impairment.
High scores on HADS (Hospital Anxiety and Depression Scale) and NQ indicate greater impairment.

Table 3 Breathing problems and the effects of stress, impact on daily life and on exercise measured on a visual analog scale (with 0 as not at all/never and 10 as very much/very often). All questions, except the last question, relate to the patient's experience of the last two months. Number of patients (*n*) and mean (SD).

	DB_1 ($n = 22$)	DB_2 ($n = 22$)	*p*-value	$Asthma_1$ ($n = 23$)	$Asthma_2$ ($n = 23$)	*p*-value
My breathing problems are negatively affected by stress	6.7 (2.8)	4.7 (3.4)	0.03	3.5 (2.9)	4.2 (3.3)	0.26
My breathing problems prevent me from exercising	4.9 (3.4)	2.0 (2.3)	<0.001	3.1 (2.7)	3.2 (2.8)	0.80
I experience breathing problems during exercise	5.0 (3.5)	3.5 (3.2)	0.046	4.3 (2.9)	4.1 (3.2)	0.70
I am afraid of developing breathing problems during exercise	3.0 (3.6)	2.0 (2.8)	0.17	2.1 (2.1)	2.1 (2.0)	0.89
My breathing problems affect my daily life	6.0 (2.6)	2.0 (2.7)	<0.001	2.6 (2.8)	2.8 (2.8)	0.66
I have had breathing problems during the last two months	7.3 (2.4)	2.9 (3.4)	<0.001	3.8 (2.9)	3.4 (3.1)	0.53
I have been off work for my breathing problems during the last year	1.7 (2.6)	0.2 (0.3)	0.02	1.5 (2.1)	0.8 (0.9)	0.25

DB = dysfunctional breathing, $_1$ = at baseline, $_2$ = at five-year follow-up.

criterion list was improved to 2.7, as compared with 6.9 at baseline ($p < 0.001$). At baseline all patients with DB had a score $\geq$5 (inclusion criterion) and at follow-up there were three patients with a score $\geq$5 in the DB group, i.e. fewer patients with DB had symptoms according to the DB criterion list (Fig. 2). No differences were seen over time in the asthma group regarding emergency room visits or the mean score on the DB criterion list.

Discussion

This follow-up study suggests that patients with DB had improved health-related quality of life, and they had fewer breathing problems both in daily life and in conjunction with exercise. Their breathing problems were also less affected by stress. Emergency room visits had decreased both regarding number of patients and number of visits, and most of the patients had fewer symptoms according to the DB criterion list. The only statistically significant difference found over time in the asthma group was regarding quality of health, bodily pain, which had deteriorated.

These improvements in the DB group could be related to the treatment they had received, trial involvement effects, spontaneous improvement of symptoms, or a combination of these factors. Owing to the design of this study, it cannot be known if it was the information and the breathing retraining that led to the improvements. Trial involvement effects and/or spontaneous improvement of symptoms may explain some of the improvements. However, the magnitude of the improvements, the fact that the patients had

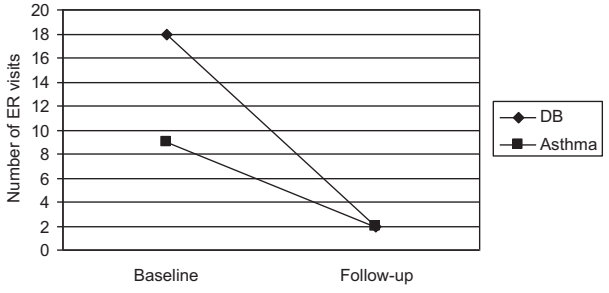

Figure 1 Number of emergency room (ER) visits for breathing problems during the last 12 months. The DB group had decreased from 18 to 2 ($p = 0.02$) and the asthma group from 9 to 2 ($p = 0.22$). DB group (numbers of patients = 22) and asthma group (number of patients = 23), at baseline and at follow-up. DB = dysfunctional breathing.

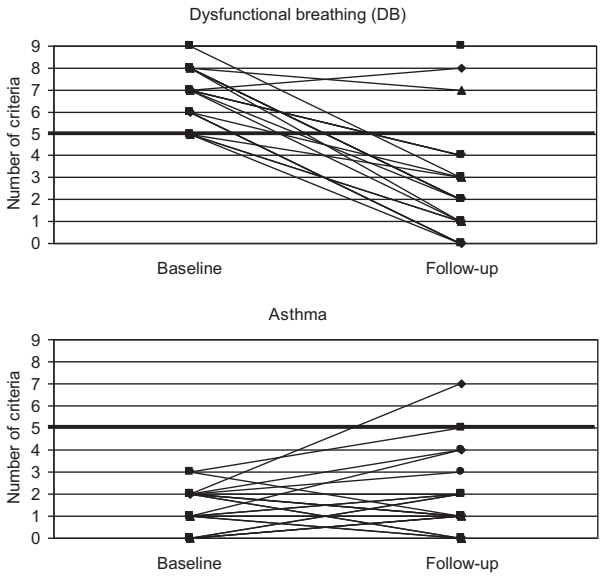

Figure 2 Results of the DB criterion list for the DB group (numbers of patients = 22) and the asthma group (number of patients = 23), at baseline and at five-year follow-up. One inclusion criterion was that patients with DB had at least five symptoms from the DB criterion list and patients with asthma fewer than five symptoms.

had their breathing problems for a mean of seven years before the intervention and the effects five years after the intervention make it unlikely this could be the full explanation. There are virtually no data on the natural course of DB and we therefore cannot rule out that the improvement in the DB group may have been spontaneous. A study on patients with sensory hyper-reactivity (SHR) a disorder that shares some similarities with DB, indicates that SHR does not improve over time.[17] Whether this also is true for DB is not known.

The patients with DB had improved in physical health (from SF-36) and they were also less impaired by breathing problems during exercise (from VAS). It might be difficult for patients to identify the reasons for their breathing problems, i.e. whether they are due to exertion, airway obstruction or abnormal breathing patterns. When symptoms remain unexplained fear of illness may become dominant[18] and it may prevent the patients from exercising. The fact that the patients with DB had received information and explanations about their condition may be a contributing factor to the improvements regarding their physical activity. It is of great value to be able to exercise, as physical activity is an important factor in reducing the risk of chronic diseases and disabilities.[19]

There were no improvements in anxiety or depression either in patients with DB or in patients with asthma in our study. Thus the decrease of respiratory symptoms in the DB group is not assumed to be attributed to improvements in anxiety and depression levels. Studies of patients with asthma have shown improvements in anxiety and depression after breathing retraining.[20,21] Delgado has shown that sense of coherence is not related to the physical measures of symptom severity from the pulmonary function tests, but to the subjective ratings of breathlessness.[22] Although the sense of coherence was stable over time in this study, the patients with DB had decreased their frequency of breathing problems.

The Nijmegen Symptoms Questionnaire cannot be used alone to diagnose hyperventilation syndrome,[16,23] although it can be used as a screening instrument.[16] It can be used as a quantitative instrument for evaluating the effects of therapeutic interventions on complaints.[23] One limitation of the questionnaire is that several questions are related to symptoms such as shortness of breath, pain and constriction in the chest — symptoms common both to asthma and dysfunctional breathing.[24] There was a decreased mean score of the NQ in the DB group, but no significant change in the number of patients with possible hyperventilation (i.e. a score $\geq$24).

The DB group showed decreased frequencies of breathing problems and their breathing problems were less affected by stress. Patients with DB had received information about their condition, increased awareness of their breathing pattern, explanations of their symptoms, and strategies for handling their breathing problems all that which may have contributed to these improvements. The patients with DB had had breathing problems for a mean period of seven years and at baseline they had had several emergency room visits for breathing problems during the last 12 months. These results are in accordance with the findings of other investigators, who showed that having breathing problems but not being diagnosed and/or not receiving optimal treatment can increase the health care consumption.[25] When symptoms remain unexplained, fear of illness may become dominant,[18] and this could lead to increased emergency room visits. Approach to treatment, to giving the patient insight into the mechanism of his symptoms can help break the vicious circle.[18] At follow-up, the emergency room visits had decreased extensively both regarding number of patients and number of visits. There was also a reduction of symptoms at follow-up, as there was a decreased score in the DB criterion list.

Breathing retraining for dysfunctional breathing in asthma has been studied[9] as has breathing retraining for asthma.[20,21] In these studies, the physiotherapy-led breathing retraining included information, diaphragmatic breathing exercises and elimination of dysfunctional breathing. The results showed improvements in quality of life and reduction of symptoms, but no effect on objective measures of respiratory function. Our study is in line with other investigations as the intervention had similarities and there was improvement in quality of life and reduction of symptoms.

One of the limitations of this study was the small sample size. However, this study may be considered a pilot study in order to access important parameters for future randomized controlled studies. Another was the fact that we did not measure carbon dioxide, because a diagnosis of hyperventilation cannot be confirmed on the basis of the questionnaire alone.[16,23] Therefore, this study does not show if some of the patients with DB really had hyperventilation with disturbed blood gases. Another disadvantage was that the DB diagnosis is not yet generally recognized and there is no existing consensus about a definition of or a diagnostic test for DB. Therefore, a 10-criteria list was developed for the baseline study.[11] It was drawn up by asking four specialists in allergology and one physiotherapist with experience of patients with DB, to independently identify the ten most common symptoms in patients with dysfunctional breathing. These symptoms provided the basis for the 10-criteria list, which was retrospectively validated against 51 medical records in patients with a high costal breathing pattern and breathing problems of a dysfunctional nature. Agreement between the 10-criteria list and the medical records was high. There was clinical consensus in the group that five out of ten criteria gave a reasonable diagnostic level. The study does not give us information about possible changes in breathing pattern five years after the intervention, as the follow-up was based on posted questionnaires.

Fourteen of the patients with DB were prescribed asthma medication before inclusion. At follow-up none of them was using asthma medication or had used asthma medication during the five-year period. Our results are in accordance with the findings of other investigators, who have shown that asthma can be overdiagnosed[1,2] and that some of these patients instead suffer from "dysfunctional breathing disorders".[2] It is important to identify and diagnose patients with DB and it is a condition to be aware of when examining patients with respiratory symptoms. Incorrect treatment due to misdiagnosis will have consequences for the patients and for health economics, i.e. adverse events attributable to ineffective asthma medication, the cost of asthma medications and emergency room

visits. DB can also occur in association with genuine asthma and other airway diseases and there is a risk that these patients are prescribed excessive doses of asthma medication.

There were substantial improvements in the DB group both in well known and validated instruments like the SF-36, HADS and NQ and in the instruments made especially for this study i.e. "Influence on daily life" (measured on a VAS) and the "DB criterion list". Values for minimal clinical changes have been estimated for the PCS (SF-36)[26] and VAS[27] in other therapeutic areas and these values were achieved in our study. However, it has not been investigated whether these values of minimal clinical changes apply to patients with DB.

This long-time follow-up study indicates that patients with dysfunctional breathing benefit from learning about their condition and from breathing retraining. As this is an observational study, the results should be confirmed by randomized controlled trials.

Authorship, contributions

Carina Hagman: designed research/study, performed research/study, collected data, analyzed data, wrote paper.

Christer Janson: contributed with important comments, analyzed data, wrote paper.

Margareta Emtner: designed research/study, contributed with important comments, analyzed data, wrote paper.

Conflict of interest statement

None of the authors has a conflict of interest to declare in relation to this work.

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